Care Coordination for Children With Sickle Cell Disease

A Longitudinal Study of Parent Perspectives and Acute Care Utilization

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Introduction: Care coordination (CC), a core element of the medical home, has the potential to reduce fragmented care and improve patient experience for children with sickle cell disease (SCD). This study aimed to (1) assess CC for pediatric SCD and (2) determine its association with acute care utilization—emergency department encounters and hospitalizations. It was hypothesized that CC would reduce acute care utilization.

Methods: A longitudinal study of 101 children with SCD was conducted. Parents completed a survey instrument on enrollment. Utilization chart review was conducted 9 months post survey. Outcome variables were emergency department encounters and hospitalizations. Independent variables were parent-reported CC, satisfaction with communication between healthcare providers, and satisfaction with communication between healthcare providers and non-medical providers (e.g., schools, child care centers). Multivariate negative binomial regression was conducted to assess associations between CC and acute care utilization. Data were collected in 2011–2013 and analyzed in 2015.

Results: One third of children had emergency department encounters and 30% had hospitalizations. At enrollment, 25% of parents reported receiving CC help and 20% reported need for extra CC. Most parents were satisfied with communication between physicians but only two thirds were satisfied with communication between their healthcare providers and non-medical providers. No significant associations were found between CC measures and acute care utilization.

Conclusions: Although parents report multiple CC deficiencies, no associations were found between CC and acute care utilization. Population-based studies are warranted to more definitively determine the association between CC and acute care utilization for children with SCD.

Introduction

The care of children with sickle cell disease (SCD) in the U.S. is marked by acute and chronic complications, including vaso-occlusive pain crises, acute chest syndrome, stroke, cognitive impairment, and organ damage.[1,2] Despite advances in care delivery and long-term survival,[3] frequent vaso-occlusive pain crises, fever, and comorbidities lead to emergency department (ED) visits and hospitalizations among children with SCD.[4–8] In a population-based study of Healthcare Cost and Utilization Project data that assessed acute care utilization among patients with SCD, the annual acute care utilization rate was 1.50 visits for children aged 1–9 years and 2.04 visits for children aged 10–17 years.[9] In a 2-year, single-site analysis of ED visits, children with SCD made 3.2 visits per year.[10] Additionally, children with SCD have higher rates of readmission relative to those with other chronic conditions, including asthma and seizure disorder.[11]

Managing SCD frequently in high-acuity settings may have negative consequences both for the individual and the healthcare system. First, providers unfamiliar with

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these children may order unnecessary tests or provide management inconsistent with the patient's long-term clinical needs. Furthermore, clinical encounters in high-acuity settings can generate substantial medical expenditures.[12] Although the chronic nature of SCD may render some of these visits unavoidable,[10,13] there has been greater focus on models of care that may reduce preventable acute care utilization.[14–17] Such models may take into account unique aspects of SCD that may impact utilization, including a high prevalence of poverty among a largely racial/ethnic minority population.[18]

The patient-centered medical home model has become a standard in health care.[19–21] As defined by the American Academy of Pediatrics, care in a medical home is accessible, continuous, comprehensive, family-centered, coordinated, compassionate, and culturally effective.[16] Although all criteria are important, increasing focus has turned to understanding the impact of care coordination (CC) on healthcare utilization.[14] CC is defined as the "deliberate organization of patient care activities between $\geq$2 participants (including the patient) involved in a patient's care to facilitate the appropriate delivery of healthcare services."[15] Within the CC framework, the comprehensive needs of the patients are met through cross-system collaborations among multidisciplinary stakeholders who may include pediatricians, hematologists, other clinicians, and social workers. According to an American Academy of Pediatrics report, CC decreases healthcare costs, reduces fragmented care, and improves the patient/family experience.[15]

Although CC has been shown to improve outcomes among children with special healthcare needs,[22,23] little is known about the impact of CC specifically among children with SCD. This study aimed to (1) determine the extent to which parents of children with SCD report CC and (2) assess the longitudinal association between parent report of CC and documented acute care utilization. It was hypothesized that children with SCD who reported to have experienced CC would have lower rates of ED visits and hospitalizations.

Methods

A longitudinal study among families of children with SCD was conducted at an urban children's hospital that follows a pediatric SCD population of 950 children annually. The hematology clinic provides both routine and urgent care services during the day. At the time of the study, CC was provided by the hematology team with no formal CC program. A survey instrument was utilized to collect demographic information and parent-reported perceptions of CC. Healthcare utilization was assessed via electronic medical record (EMR) review for the 9-month period following enrollment. EMR review was conducted manually and verified with EMR data extrapolations by a system analyst. Written informed consent from parents and child assent waivers were obtained for all study participants. The study was approved by the IRB of Baylor College of Medicine, Houston, Texas.

Study Population

Study participants were recruited during outpatient hematology clinic visits or while hospitalized at Texas Children's Hospital. Subjects were parents or guardians (hereafter caregivers) of children aged 0–17 years. Eligible caregivers were those who had children with ICD-9-CM codes consistent with hemoglobin SS disease, sickle hemoglobin C disease, or sickle beta zero thalassemia. Caregivers unable to comprehend English were excluded from the study.

All potential participants were systematically screened for eligibility prior to recruitment. An attempt to recruit all eligible participants for the study was made. Prospective caregivers were initially informed of the study by their child's provider. Those interested in participating were recruited and consented by a research coordinator. Caregivers were asked to complete a survey instrument assessing their CC experience and whether or not they could have used additional assistance. Caregivers provided consent for 9-month prospective review of their child's records. Participant recruitment took place from September 1, 2011, through September 5, 2012. Data collection concluded on June 5, 2013, 9 months after the last study participants enrolled.

Measures

The primary outcome variables were EMR-documented ED encounters and hospitalizations obtained from 9-month prospective chart review. An ED encounter was defined as an ED visit that resulted in discharge from the ED without hospital admission. Therefore, there was no overlap between ED encounter and hospitalization outcomes.

The primary independent variables were individual components of CC as measured in the survey instrument: (1) caregiver-reported help with CC; (2) satisfaction with communication between healthcare providers; and (3) satisfaction with providers' communication with non-medical service providers (e.g., school, child care centers). The survey instrument administered to caregivers consisted of questions about the child, family, and receipt of care. The survey included questions regarding experiences with different components of CC, all of which were taken verbatim from the 2007 National Survey of Children's Health (NSCH).[24] Though other measures of CC exist,[25] the NSCH was selected for several reasons. First, it examines multiple aspects of the CC concept, allowing evaluation of a wide spectrum of CC functions. Second, numerous population-based studies have used this measure,[26–28] providing the opportunity to compare findings. Lastly, the NSCH definition of the patient-centered medical home, including CC, has been endorsed by the National Quality Forum.[29]

Child covariates consisted of gender; age; SCD genotype; caregiver-reported health status; insurance type; number of office visits (during the 9-month enrollment period); and hydroxyurea usage (during the 9-month enrollment period). Insurance type was categorized as public, private, or uninsured. To assess health status, the NSCH item asking caregivers *In general, how would you describe [CHILD'S NAME]'s health?* was used. Caregiver options

consisted of *excellent*, *very good*, *good*, *fair*, or *poor*. For the purpose of this study, responses were categorized as *excellent/very good/good* versus *fair/poor*. Caregiver covariates consisted of age, gender, relationship to child, marital status, education level, and number of additional children in the household.

Statistical Analysis

Summary statistics were used to determine the percentage of children achieving specific components of CC. Multivariate negative binomial regression analyses, which can correct for overdispersed count data, were used to assess associations between CC items and acute care utilization (ED encounters, hospitalization). The relationships between CC and acute care utilization were assessed in two different models for each outcome. The models differed on how CC was defined as an independent variable. The first model included the individual measures of care coordination as independent variables. In the second model, ordinal specification was used where the independent variable consisted of the number of CC components achieved (0, 1, 2, 3). The CC items included in this variable consisted of anyone helping to coordinate care (yes versus no); satisfaction with communication between providers (very satisfied/somewhat satisfied versus very dissatisfied/somewhat dissatisfied); and satisfaction between provider and non-medical service providers (very satisfied/somewhat satisfied versus very dissatisfied/somewhat dissatisfied). Results were calculated as incident rate ratios for the number of encounters per child with 95% CIs. All analyses were controlled for child and caregiver covariables. Statistical analyses were performed in 2015 using SAS, version 9.2.

Results

Of 151 caregivers approached for study participation, 101 were enrolled and completed the survey instrument, yielding a participation rate of 67%. Seven caregivers declined participation citing lack of time or desire to consult other family members. The remaining 43 caregivers approached expressed interest in the study but never completed the survey instrument.

The sociodemographic characteristics of the study sample (N=101) are shown in Table 1. All subjects were African American. Boys accounted for half of the sample of children. The mean child's age was 7.8 years. Eighty-three percent of children were reported to have excellent/very good/good health status. Approximately three quarters of the children were publicly insured. Children attended an average of 4.8 hematology office visits in the 9 months following enrollment. A total of 47 (46.5%) children were prescribed hydroxyurea during the study period.

In terms of respondent characteristics, 96% of respondents reported to be the child's parent and 30% were married. More than 97% of respondents were female. Nearly three quarters of respondents had other children in the household.

Table 1. Patient and Family Characteristics Among Children With Sickle Cell Disease Treated at an Urban Children's Hospital

Characteristics	Overall, *n* (%) (N=101)
Patient characteristics	
Gender	
Male	47 (46.5)
Age (years)	
0–4	22 (21.8)
5–12	64 (63.4)
13–17	15 (14.9)
Genotype	
SS	88 (87.1)
SC	8 (7.9)
Sβ0thal	5 (5.0)
Health status	
Excellent/very good/good	84 (83.2)
Fair/poor	17 (16.8)
Insurance	
Public	74 (73.3)
Private	24 (23.8)
Uninsured	2 (2.0)
Both	1 (1.0)
Number of office visits	
M (SD)	4.8 (3.2)
Median (range)	4 (0–18)
Prescribed hydroxyurea	47 (46.5)
Caregiver characteristics	
Gender	
Female	98 (97.0)
Age (years)[a]	
20–30	26 (25.7)
31–40	55 (54.5)
41–50	14 (13.9)
≥51	5 (5.0)
Relationship to child	
Parent	97 (96.0)
Grandparent	2 (2.0)
Step-parent	2 (2.0)

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Table 1. Patient and Family Characteristics Among Children With Sickle Cell Disease Treated at an Urban Children's Hospital *(continued)*

Characteristics	Overall, *n* (%) (N=101)
Education[a]	
High school or less	26 (25.7)
Some college	42 (42.4)
College degrees	31 (31.3)
Marital status	
Not married	71 (70.3)
Married	30 (29.7)
Other children in household[a]	
None	21 (20.8)
One	38 (37.6)
Two or more	37 (36.6)

[a]Individual categories may not add up to 101 because of missing responses.

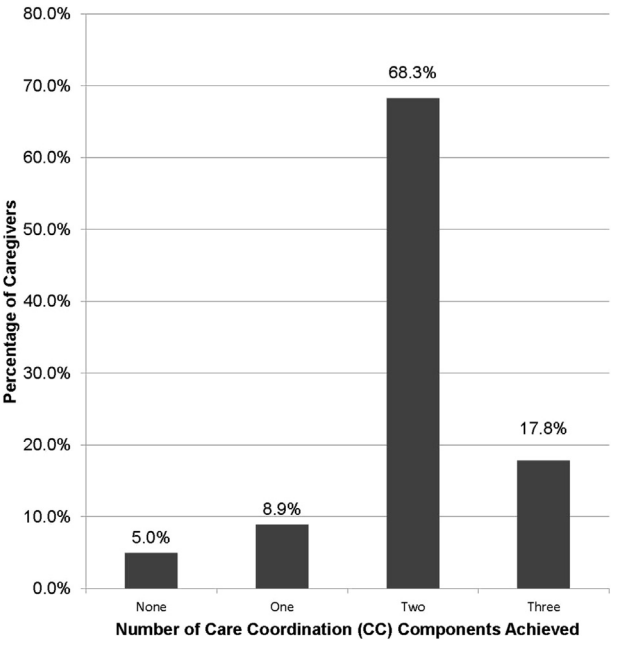

Figure 1. Components of care coordination achieved among children with sickle cell disease.
Note: Components of CC: Received help coordinating care, Satisfied with communication among providers, Satisfied with communication between doctors and non-medical service provider.

In total, 87% (*n*=88) of caregivers reported having a primary care pediatrician for their child. The receipt of CC among the sample is shown in Table 2. Only 25% of caregivers reported that someone helped them coordinate their child's care, and 18% reported that they could use extra help coordinating care. Caregiver satisfaction with communication between providers was 88% whereas reported satisfaction with provider communication with non-medical providers was 21% lower. Figure 1 demonstrates the number of CC components achieved—someone helping coordinate care (yes); satisfaction with communication between providers (very satisfied/ somewhat satisfied); and satisfaction with communication between provider and school and community (very satisfied/somewhat satisfied). Although 95% reported receiving at least one component of CC, only 17.8% received all three CC components.

Analysis of ED use showed that one third (*n*=34) of children had a total of 60 ED encounters. Within this group, the median number of ED visits per child was 1.7 (range, 1–6). Two thirds of children (*n*=67) had no ED visits. A total of 31 children were hospitalized, resulting in 58 inpatient stays. The median number of hospitalizations per hospitalized child was 1.8 (range, 1–9). Twelve percent of children (*n*=12) had both ED encounters and hospitalizations. Forty-eight percent (*n*=48) of children had no hospitalizations. The most common diagnoses across encounter types were pain, vaso-occlusive pain crises, and acute chest syndrome.

For multivariate analyses, only respondents who completed all survey items (*n*=88, 87%) were included. No associations were found between individual CC components and ED encounters or hospitalizations. In addition to individual CC components, associations between number of CC components achieved and acute care utilization were also assessed. No associations were found between number of CC components achieved and ED encounters or hospitalizations.

Table 2. Caregiver Self-Reported Receipt of Care Coordination in an Urban Children's Hospital (N=101)

Component	*n* (%)
Received help coordinating care	25 (24.8)
Satisfied with communication among providers	89 (88.1)
Satisfied with communication between doctors and non-medical service provider	68 (67.3)

Discussion

In this study of children with SCD cared for in an urban children's hospital, a comprehensive assessment of CC is provided. Although almost 90% of children were reported to have a regular provider, they experienced multiple

deficiencies in CC. More specifically, less than 25% of caregivers reported having someone to help them coordinate their child's care and nearly 20% reported needing extra help coordinating care.

A number of studies provide context for the findings. In a population-based study by Toomey et al.[26] using the NSCH, 31% of children were found to have unmet CC needs and 40% of children with special healthcare needs were reported to have unmet CC. In that study, effective CC was defined as a composite variable consisting of three components: (1) getting as much help as needed with coordinating care; (2) caregiver satisfaction with communication among healthcare providers; and (3) caregiver satisfaction with communication between healthcare providers and non-medical service providers. In a study using the National Survey of Children with Special Health Care Needs, Turchi and colleagues[22] found that 68.2% of families received some type of CC; however, 40.8% of those families reported inadequate CC. In contrast to these studies, the current study assessed specific items rather than a composite variable, given the goal of identifying specific deficiencies. Specific deficiencies were found in needs for extra CC and satisfaction with communication between healthcare provider and non-medical providers. This study builds on previous work in assessing CC within a specific subgroup of children.

Although multiple deficiencies in CC were reported, no statistically significant associations were found between CC and acute care utilization. The lack of association between CC and acute care may be attributable to several factors. First, it is possible that CC does not impact acute care utilization. Children with chronic conditions may have greater levels of illness severity and therefore require acute care encounters. Although some encounters may be preventable, others may not be, especially those occurring at off hours. Prior studies have shown limited associations between components of the medical home and acute care utilization.[30–32] Second, it is possible that the definition of CC in the NSCH may not be applicable across all populations of children. Therefore, these CC components would not impact acute care utilization for children with SCD.

This study raises several issues about CC as it pertains to children with SCD. The questions from the NSCH allow for comparisons across studies but may not be specific enough to the SCD population. For children with SCD, attention needs to be directed toward CC beyond the medical system, as CC addresses the "social, developmental, educational, and financial needs of patients and family."[15] In this study, only 69% of caregivers of children with SCD were satisfied with the communication between doctors and non-medical providers, including schools and child care centers. This is a major issue for families with children with SCD. In this study, children with SCD attended an average of 4.8 office visits in a year. Similarly, in a study by Schwartz et al.,[33] adolescents with SCD attended an average of 4.5 routine office visits in a year. These numbers only account for routine hematology visits and do not include encounters for chronic transfusions, primary care, or other specialists who manage comorbidities. These appointments, which are typically during school hours, can result in missed classroom time and undermine school attendance. In addition, as a result of sociodemographic factors, strokes, anemia, and cognitive impairments, children with SCD have been shown to have poor educational attainment.[34] Therefore, measures of CC for children with SCD may require more focus on cross-system collaboration with schools.

Although assessment of CC was the major focus of this study, a related issue is how to actually provide effective CC to children with SCD given challenges inherent to this population. First, although screening tools are in development,[35] there is no standard method in clinical care settings for identifying children in need of CC. Individual children with SCD may require different components or levels of CC at different points in time. Second, CC for a child depends on engagement of caregivers. Parents of children with chronic diseases, including SCD, have been shown to be less involved in disease management (e.g., appointments, medication management) during adolescent years.[36] Third, payment for CC has had limited success as the cost of CC is not directly reimbursable under many traditional payment models.[15,37,38] However, recent inclusion of CC in the Current Procedural Terminology Manual may increase opportunities for adequate reimbursement through both private and public insurance.[15]

For children with SCD, the challenges of CC are compounded by structural barriers, including limited communication between clinical providers, limited communication between clinical providers and schools, inadequate insurance coverage, poverty, cultural barriers, and discrimination.[18,39,40] CC interventions designed for pediatric SCD must address the unique barriers of this population.

Limitations

This study had several strengths, including a comprehensive assessment of CC, longitudinal design, and use of EMR-documented healthcare utilization. However, several limitations of the study warrant discussion. Although endorsed by the National Quality Forum as a component of an overall medical home measure, the assessment of

CC used in this study differs from other measures both in terms of assessed components and validation of its contents.[25] Currently, no gold standard measure of CC exists. Information used to determine whether a child experienced CC was derived from caregiver report rather than clinical sources. Therefore, the data were subject to recall bias. In conducting negative binomial regression with ordinal specification, it is assumed that all components of CC had equal value. However, it is possible that certain components may be more important than others. For example, active collaboration between healthcare providers may especially impact unnecessary healthcare utilization through reduction of care fragmentation.[14] Generalizability was limited by this being a single site study. Children cared for in an urban children's hospital may be fundamentally different from other SCD populations. The sample size was small relative to national studies assessing CC among children. Therefore, the authors could not detect small differences between groups and variables. Lastly, because caregivers were not asked to specify which provider served as the reference for their survey responses, it could not be determined whether caregivers were responding to questions based on their primary care experiences or those with their hematologist. This may have limited ability in determining where specific CC gaps existed. However, as CC represents functions of care across a network of providers and locations, the identified deficiencies may reflect gaps in co-management of care between primary care and hematology rather than in just one specific setting.

Conclusions

As CC is increasingly promoted as an integral function of healthcare services for children with chronic conditions, data are starting to emerge on how it fulfills the "triple aim" of health system transformation consisting of improved population health, reduced healthcare costs, and enhanced patient experience.[41] However, little is known regarding to what extent children with SCD experience CC and whether CC can impact health outcomes in this population. These findings demonstrate that children with SCD experience multiple deficiencies in CC. However, among the sample in this longitudinal study, no associations were found between CC acute care utilization. Future studies should include multiple centers and be population-based. Subsequent studies should also assess CC functions more specific to SCD and a wider spectrum of behavioral, educational, and health outcomes, including quality of life[42] and school and family functioning.

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TLR conceptualized the concept and design of the study; acquired, analyzed, and interpreted the data; drafted the initial manuscript; and critically revised the manuscript for important intellectual content. AEW and HF performed the statistical analysis in the study and provided administrative, technical, and material support. JLR conceptualized the concept and design of the study, analyzed and interpreted the data, and critically revised the manuscript for important intellectual content.

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